

Unprecedented Remission with Radiotherapy Alone: A Rare Case of Mutilating Scalp Recurrence of Hodgkin's Lymphoma with Preexisting Chronic Liver Disease

Abstract

Recurrence of Hodgkin's lymphoma in the scalp is extremely rare (only one such case reported, way back in 1970) and its treatment in the setting of chronic liver disease is unknown. We hereby report the second case of recurrent Hodgkin's lymphoma of the scalp, in a 28-year-old male, presenting as a mutilating ulcer over the entire right hemicranium with autoamputation of the right pinna. He was unfit for chemotherapy in view of his preexisting chronic liver disease and was therefore treated with radiotherapy alone, to the dose of 40 Gy in 20 fractions over 4 weeks in conventional fractionation, using Tele-Cobalt 60, leading to complete and sustained remission (at 5 years follow up) and unprecedented organogenesis of the scalp and the pinna.

Keywords: Complete remission of mutilating ulcer, Hodgkin's lymphoma of the scalp, Hodgkin's lymphoma recurrence, organogenesis, radiotherapy alone

Introduction

The scalp, as a site for lymphoma, is mostly primary and of non-Hodgkin's lymphoma (NHL) histology, but Hodgkin's lymphoma of the scalp in the primary setting has not been reported in literature. Till date, only one case of recurrent Hodgkin's lymphoma in the scalp had been reported, way back in 1970, in a patient with no other comorbidities.

We hereby report the second case of its kind, a recurrent Hodgkin's lymphoma of the scalp (site of previous disease in the preauricular, cervical, portal, and retroperitoneal lymph nodes), presenting as a mutilating ulcer and autoamputation of the pinna in a young male, who was unfit for chemotherapy due to chronic liver disease, and subsequently, underwent complete and sustained remission with radiotherapy alone and showed unprecedented organogenesis.

Case Report

A 28-year-old male, who was being treated for a chronic hepatitis C in the School of Digestive and Liver Disease of our super specialty centre IPGMER and SSKM Hospitals, Kolkata, was referred to the Department of Radiotherapy in July 2012, with a painless nonhealing ulcer over the

scalp, at the right temporal region measuring 15 cm (A-P) × 10 cm (S-I) × 1 cm (depth).

The history of the patient revealed that, in August 2008, he was diagnosed and treated for Hodgkin's lymphoma Stage IIIA in another center which is a nonsuper-specialty hospital. He presented there with a progressive swelling over the right parotid region for 6 years, without any B symptoms. The papers revealed clinical and radiological evaluation reports of a large lymph nodal mass palpable over the right parotid region (preauricular lymphadenopathy) along with right Level IB and II cervical lymphadenopathy, hepatosplenomegaly, portal, and retroperitoneal lymphadenopathy. Incision biopsy reported Hodgkin's lymphoma with no further subtyping or immunohistochemistry (IHC), serum lactate dehydrogenase (LDH) was 401 U/L, and bone marrow was not done. He was treated with four cycles of Adriamycin 25mg/m², Bleomycin 10 IU/m², Vinblastin 6mg/m², Dacarbazine 375mg/m² (ABVD) followed by four cycles of COPP (Cyclophosphamide, Vincristine, Procarbazine and Prednisolone in their standard doses), followed by mantle field irradiation 40 Gy in 20# (completed on June 05, 2009), with no detailed reasoning for the choices of the treatment done. He

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was not under any follow-up in the primary treating hospital, because he was diagnosed with hepatitis C virus (HCV) infection within months after treatment completion for his lymphoma, and was referred to the school of liver disease in the current institute for treatment. He was receiving treatment for his liver disease in this institute since 2010.

The current ulcer started as a small nodule in January 2012 and progressively grew in size over 2 months destroying the entire scalp over the right hemicranium, including near autoamputation of the right pinna [Figure 1] showing the non healing ulcer at presentation. Kindly appreciate the white gauze placed in the tissue lost area of the right pinna, as evident in the image provided as Figure 1.

Biopsy from the ulcer margins showed features suggestive of Hodgkin's lymphoma. [Figure 2a and b] Tissue covered by stratified squamous epithelium with hyperplasia. Sub-epithelial region is composed of sheets of cells with a polymorphic appearance - lymphocytes, histiocytes, eosinophils and plasma cells. Hodgkin's cells and Reed Sternberg like cells are present. IHC from the block show R-S cells expressing CD 30, CD 20, and Pax-5

and negative for leukocyte common antigen, CD 15, and CD 3. The background was rich in T cells. Fluorescence *in situ* hybridization for Epstein-Barr encoding region was negative (IHC images could not be traced from the department of Pathology). Finally, the disease was classified as a case of recurrent Classical Hodgkin's lymphoma of the scalp (further subtyping not provided). Contrast-enhanced computed tomography of the neck, thorax, and abdomen, and bone marrow trephine biopsy revealed no evidence of the disease, except for hepatosplenomegaly, raised liver enzymes (aspartate aminotransferase/alanine transaminase: 86/142 mg/dl), and serum LDH 505 U/L.

In view of his chronic liver disease, he was treated with external beam radiotherapy alone, to a dose of 40 Gy in 20 fractions over 4 weeks, using medial, and lateral oblique tangential beam technique, with wet gauge bolus, using the ATC-C9 cobalt 60 machine.

On response evaluation, the ulcer started to heal within the first 2 weeks after the first 20 Gy of radiation was completed. [Figure 3] Response after 20 Gy in 10 fractions of radiotherapy. The 3rd week marked the beginning of the organogenesis of the right pinna. By 3 weeks after radiotherapy, the lesion was partially covered with skin and rest with granulation tissue and the pinna was nearly



Figure 1: The nonhealing ulcer at presentation

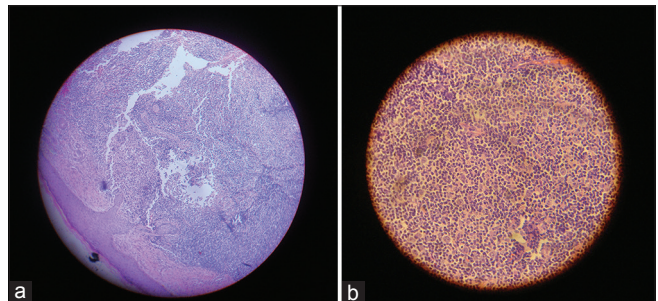


Figure 2: (a and b) Tissue covered by stratified squamous epithelium with hyperplasia. Subepithelial region is composed of sheets of cells with a polymorphic appearance - lymphocytes, histiocytes, eosinophils, and plasma cells. Hodgkin's cells and Reed-Sternberg-like cells are present



Figure 3: Response after 20 Gy in 10 fractions of radiotherapy



Figure 4: Response at 4-month follow-up after radiotherapy

completed in shape. At the 6th week of follow-up, the lesion was 90% covered with skin and remaining with granulation tissue. On the 4th month of follow-up, the lesion was completely covered with skin and the pinna was in a normal shape, with no evidence of any residual disease [Figure 4] Response at 4 Months Follow up after radiotherapy.

Now after 57 months of follow the patient is still in a complete remission from the Hodgkin's lymphoma [Figure 5]. Response at 57 months follow up after radiotherapy. He developed a small raw area because of an itch which was biopsied to be free from any residue or recurrence. The itch and the raw area subsided with local antifungal-antibacterial cream and oral H1 blockers. The status of his liver function and general health, though, is on the downfall.

Discussion

Relapse rates of Hodgkin's lymphoma range from 10% to 20% in Stage I–II disease and 11%–40% in patients with more advanced disease,^[1] but the sites of recurrence remain the lymph nodes, previously irradiated as well as new and lymphoid tissue.^[2] Recurrence of this disease in the scalp is extremely rare as it is neither in contiguity with other lymph nodes in the body nor is it a lymphoid organ.

Scalp lymphomas are usually primary and of NHL histology. Hodgkin's lymphoma recurring in the scalp (noncontiguous with the previous site of disease) had hardly been reported in literature. On detailed review of literature, only one such case was reported in an article published in 1970, from Downstate Medical Centre in Brooklyn, NY, where a review of 134 patients with Hodgkin's disease was presented. It was found that only ten patients relapsed in the skin, and the common feature of all these relapses was the proximity of initial lymph nodal involvement and the subsequent skin involvement within its drainage area, suggesting a retrograde lymphatic spread. Of these ten patients, the

one that relapsed in the scalp of the forehead had an initial disease in the preauricular lymph node, exactly similar to the case reported here.^[3]

Epidemiological studies done over 20 years showed an association of HCV infection with diffuse large B-cell lymphoma.^[4] In Asian population with HCV infection, such a risk is twice as high as compared to the normal population. However, no such association is known to exist between HCV infection and Hodgkin's disease.^[5]

This patient who had presented with a recurrent Hodgkin's lymphoma as a mutilating ulcer over the scalp with near autoamputation in the back drop of chronic liver disease left us with no choice other than a trial of radiotherapy to the scalp.

The overwhelming results with complete and sustained remission and unprecedented organogenesis, as seen in this case, shows that scalp recurrence of Hodgkin's lymphoma may be cured with radiotherapy alone without any need for systemic treatment. The outcome achieved in this case is exactly similar to that of the only patient previously reported.^[3]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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Figure 5: Response at 57-month follow-up after radiotherapy